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Thoracoscopic treatment of a broncho-esophageal fistula: A case report

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ABSTRACT

INTRODUCTION: Broncho-esophageal fistula is a rare clinical condition which can be manifested with non-specific signs and symptoms.**PRESENTATION OF A CASE:** Here, we report an adult case of a broncho-esophageal fistula in a 43-year-old man referred for chronic cough after fluid food intake and weight loss. Barium swallow, esophagogastroduodenoscopy, bronchoscopy and Computed Tomography of the chest demonstrated a broncho-esophageal fistula between the apical segmental bronchus of the lower right lobe and the middle section of the esophagus. The patient underwent video-assisted thoracoscopic surgery for resection of the fistula. No post-operative complications occurred.**DISCUSSION:** Broncho-esophageal fistula in adults is rare and its diagnosis is often delayed due to the frequent lack of specific symptoms. Although there is no standard protocol, the most widely used treatment is thoracotomy with identification and dissection of the fistula tract followed by repair of bronchial and esophageal defects.**CONCLUSIONS:** Video-assisted thoracoscopic surgery appears to be an effective and minimally invasive approach for the treatment of broncho-esophageal fistulas, especially in young, healthy subjects.© 2016 The Authors. Published by Elsevier Ltd on behalf of IJS Publishing Group Ltd. This is an open access article under the CC BY-NC-ND license (<http://creativecommons.org/licenses/by-nc-nd/4.0/>).

1. Introduction

Broncho-esophageal fistula (BEF) is uncommon in adult subjects. It may be congenital or acquired. Congenital BEF is most frequent in infancy, when it is usually due to non-completion of the tracheoesophageal division in the early stage of embryonic development [1,3]. Acquired BEF may derive from malignant pathologies such as esophageal, bronchial or mediastinal neoplasms. Benign types are less common, with that of tubercular origin being the most frequent for this category. Remaining cases of acquired BEF are of a benign nature as a result of trauma or infection. Traumatic forms include iatrogenic BEF as well as those secondary to the ingestion of foreign bodies or harmful substances.

Undiagnosed congenital BEF may not result in complications until adulthood, depending on its size and location, or may cause non-specific signs and symptoms such as recurrent episodes of pneumonia, pulmonary abscess, chest pain, hemoptysis and low-

grade fever [4,5], although coughing following oral fluid intake (Ono's sign) is present in approximately 65% of cases [6]. These symptoms can be present for 6 months to 50 years before fistula is diagnosed [7]. They are more frequently diagnosed when the subjects are in their thirties, with no difference in frequency between the sexes [8]. Late manifestation of this congenital anomaly could be explained with multiple mechanisms. They include the oblique course of the fistula, the presence of a membrane blocking the lumen of the fistula, the presence of a valve mechanism and spasm of the fistulous tract [9]. Nevertheless, in many instances in adulthood it is difficult to establish the real congenital or acquired origin of a BEF.

There are four types of BEF as classified by Braimbridge and Keith [10]. Type 2 is the most common (almost 90%), and is due to having a short tract running directly from the esophagus to the bronchus.

Broncho-esophageal fistula requires surgical repair [5] and it should be performed as soon as the diagnosis is established. The surgical approach is usually by thoracotomy. The VATS approach to BEF seems to be an effective approach without significant morbidity, and quickly resolves the pathology. During the diagnostic work-up an x-ray of the upper digestive tract with water-soluble contrast is highly specific, minimally invasive, and more easily performed. Further examinations that may prove helpful are esophagogastroduodenoscopy and bronchoscopy, which may reveal the

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opening of the fistula tract. We present a rare case of broncho-esophageal fistula (BEF) in an adult patient with the aim of proposing a minimally invasive approach by Video-Assisted Thoracoscopic Surgery (VATS).

2. Presentation of a case

A 43-year-old man, with no history of smoking, tuberculosis, respiratory diseases or trauma presented with a persistent non-productive cough with mainly postprandial onset with duration of approximately two years. The cough was particularly irremissible after the intake of fluids. This had resulted in significant weight loss due to reduced food intake. Blood tests at presentation were normal. Computed Tomography (CT) of the thorax showed a hypodense lesion in the right pulmonary lower lobe. Water-soluble contrast radiography of the esophagus showed a broncho-esophageal fistula between the apical segmental bronchus of the lower right lobe and the middle section of the esophagus (Fig. 1a). Bronchoscopy showed a fistula opening that was 4 mm in diameter and approximately 2.5 cm distal to the main carina subsequently confirmed by CT scan 3D rendering (Fig. 1b). Esophagogastroduodenoscopy confirmed a fistula tract ending in the esophageal tract at 30 cm from the incisors. The patient underwent VATS under general anesthesia with selective ventilation and low levels of insufflation pressure in the right lung. The fistula tract was stapled at both ends using an Endo GIA device and excised. The chest tube (24 F placed through lower trocar incision) was removed on the first postoperative day. At the same time, a new esophagography was performed to show any esophageal leakage (Fig. 2). The patient resumed solid food 24 h after the operation. No post-operative complications were observed and there was complete remission of the presenting symptoms. He was discharged in good health on the third postoperative day. Two years after follow-up, there are no apparent complications.

3. Discussion

Broncho-esophageal-fistulas are rarely diagnosed in adults [11]. Although symptoms of this condition are aspecific, significant delays in its diagnosis cannot be excused. If not suspected and left

untreated, BEF may lead to fatal complications despite its benign nature. Moreover, as in our case, patients complain of symptoms that interfere considerably with their quality of life and working capacity. Currently, most studies concerning BEF are case reports, and few clinical data are available that are based on large samples. Surgical management is the fundamental point for the prognosis of patients with BEF [4]. At present, there are no standardized treatment protocols. The most frequent approach is thoracotomy with identification and section of the fistula followed by repair of the bronchial and esophageal defect [1,12]. This may sometimes necessitate additional interventions such as lobectomy or pneumonectomy. A cervical approach is also described in the literature for BEF located in the upper esophageal tract [13]. A less common alternative is the endoscopic injection of sclerosing substances (sodium hydroxide and acetic acid) at the esophageal and bronchial openings of the fistula [14]. However, this procedure seems more suitable for congenital fistulas of moderate size [15]. Endoscopic insertion of self-expandable metal stents is mainly indicated to palliate symptoms in neoplastic patients with a poor prognosis [16,17]. Another endoscopic option is the combined injection of fibrin glue by means of bronchoscopy, and the insertion of a metal clip by esophagoscopy [18,19]. We believe that VATS may be an effective and minimally invasive approach, suitable particularly in young patients in apparently good health and who tolerate homolateral pulmonary exclusion during thoracoscopic maneuvers. However, it is uncertain whether it can be used in the case of a large fistula or where adhesions make isolation of the fistula difficult. In our case, thoracoscopy was easily performed because of low adhesions of lung parenchyma and a sufficiently long fistula tract. However, we believe that the use of the thoracoscopic technique strictly depends on individual surgical experience. In conclusion, the type of the fistula is the main criteria for treatment choice on an individual basis, with the options being either thoracotomy or thoracoscopy, or, in exceptional cases, endoscopy with sclerosing materials or stents. Thoracotomy can be an option if thoracoscopy is not successful, and it can be subsequently carried out during the same intervention. Therefore, we can confirm that the rarity of this pathology and lack of surgical experience do not permit definite conclusions to be drawn regarding the best therapeutic approach; further prospective studies are necessary to better address the issue.

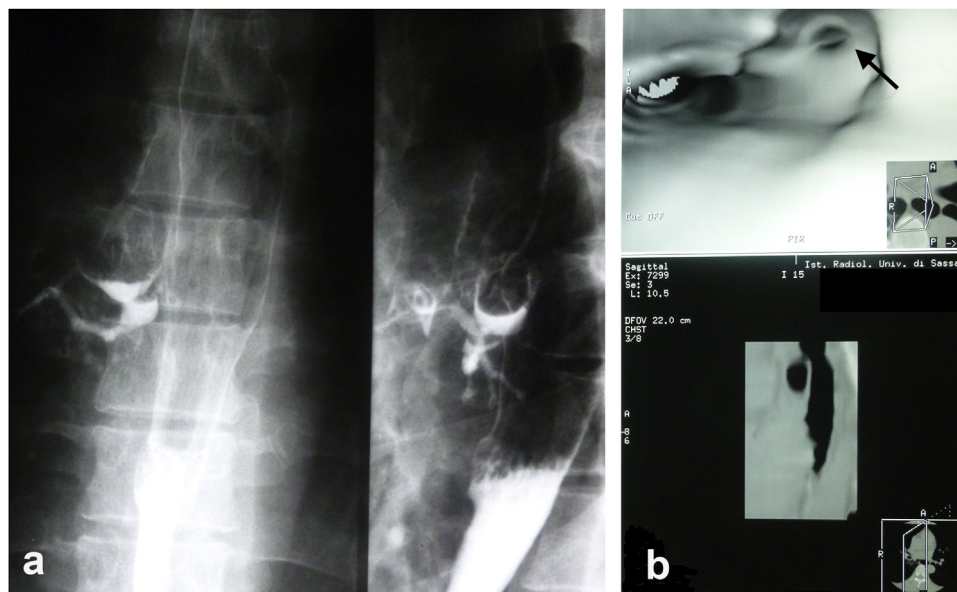


Fig. 1. (a) Broncho-esophageal fistula between the middle section of the esophagus and the apical segment bronchus of the lower right lobe demonstrated at X-ray with water-soluble contrast medium. (b) CT scan 3D rendering confirmed the diagnosis of broncho-esophageal fistula.

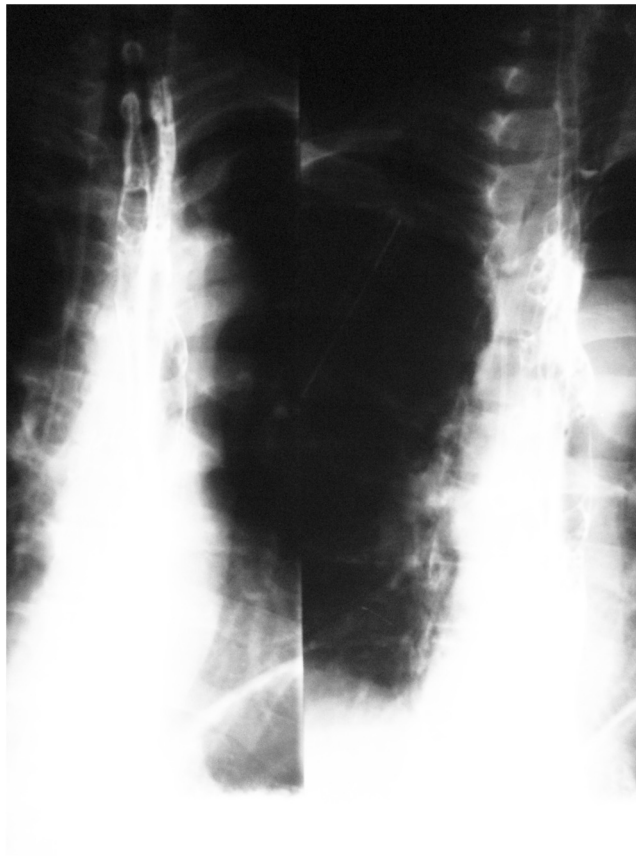


Fig. 2. X-ray of the upper digestive tract with water-soluble contrast performed the day after the operation showed no fistula tract.

Written informed consent was obtained from the patient for publication of this case report and accompanying images. A copy of the written consent is available for review by the Editor-in-Chief of this journal on request.

Conflicts of interest

All authors have no substantial direct or indirect commercial financial incentive associated with publishing the manuscript.

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Ethical approval

Whether approval by Institutional Board has been given for this case report.

Consent

Informed consent was obtained from the patient; all authors ensure that all text and images alterations to protect anonymity do not distort scientific mean of the manuscript.

Author contribution

Giorgio C. Ginesu: Writing paper.
Claudio F. Feo: data analysis.
Francesca Ruii: data analysis.
Francesca Addis: data collection.
Maria L. Cossu: Text edit.

Alessandro Fancellu: Writing paper.
Alessandro G. Fois: Text edit.
Panagiotis Paliogiannis: data collection.
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Registration of Research Studies

This section does not apply to hour case report.

Guarantor

Giorgio C Ginesu.

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